

Embolic Stroke due to Carotidynia Potentially Associated with Moving Carotid Artery Caused by Swallowing

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A 63-year-old woman with end-stage renal disease on maintenance hemodialysis discontinued her medication for rheumatoid arthritis with prednisolone and azathioprine. One month later, she was admitted because of consciousness disturbance and right hemiparesis. Diffusion-weighted brain magnetic resonance imaging (MRI) revealed multiple hyperintensities in her left frontal and parietal lobes. She also developed high fever and left neck pain. Carotid ultrasonography showed calcified plaque with vessel wall swelling at the bifurcation of the left common carotid artery (LCCA) and surrounding hypoechoic soft tissue. The tissue was identified as an isodense lesion on noncontrast computed tomography (CT) and as a high-intensity lesion on fat-saturated T2-weighted MRI. From her symptoms and radiological findings, she was diagnosed with carotidynia. Cervical MRI also showed that the LCCA was transposed to a retropharyngeal location, suggesting a moving carotid artery. Carotid ultrasonography revealed that the LCCA moved to and from the retropharyngeal position with swallowing and was thus being compressed by the hyoid bone. After corticosteroid therapy was initiated with 30 mg of prednisolone, her symptoms and radiological findings improved. To our knowledge, this is the first report of a case of cerebral embolism due to carotidynia. The repetitive compressions by the hyoid bone during swallowing were presumed to have provoked shear stress and inflammation of the carotid vessel wall, which was aggravated by discontinuation of steroid therapy in our case. These mechanical and inflammatory stresses might cause dysfunction of endothelial cells, hypercoagulation, platelet hyperaggregation, and vulnerability and rupture of carotid plaques, and may subsequently result in embolic strokes. **Key Words:** Carotid ultrasound—carotidynia—embolic stroke—hyoid bone—moving carotid artery.
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Introduction

Carotidynia, as first described by Fay in 1927, is an idiopathic neck pain syndrome characterized by radiating pain and tenderness over the common carotid bifurcation.¹ Although its etiology remains uncertain, inflammation is considered the most important cause.² Recently, specific imaging findings for the diagnosis of carotidynia using ultrasonography, computed tomography (CT), magnetic resonance imaging (MRI), and positron emission tomography have been reported.³⁻⁵ Arterial thrombosis associated with carotidynia is rare,⁶ and cerebral

embolism caused by carotidynia has not been reported. We herein report a rare case of cerebral embolism due to carotidynia.

Case Report

A 63-year-old woman with end-stage renal disease on maintenance hemodialysis discontinued her medication for rheumatoid arthritis with prednisolone and azathioprine. One month later, she was admitted because of consciousness disturbance and right hemiparesis. Diffusion-weighted brain MRI revealed multiple hyperintensities in her left frontal and parietal lobes (Fig 1, A). She also developed high fever and left neck pain. Laboratory examination revealed elevated inflammatory markers: C-reactive protein 23.2 mg/dL, erythrocyte sedimentation rate greater than 110 mm/h, serum amyloid A 417 $\mu\text{g}/\text{mL}$, and soluble intercellular adhesion molecule-1 583 ng/mL. D-dimer was also elevated at 6.1 $\mu\text{g}/\text{mL}$. Carotid ultrasonography showed a calcified plaque with vessel wall swelling at the bifurcation of the left common carotid artery (LCCA) with surrounding hypoechoic soft tissue (Fig 1, B,C). The tissue was identified as an isodense lesion on noncontrast computed tomography (CT) (Fig 1, D) and as a high-intensity lesion on fat-saturated T2-weighted MRI (Fig 1, E). From her symptoms and radiological findings, she was diagnosed with carotidynia. Cervical MRI

also showed that the LCCA was transposed to a retropharyngeal location, suggesting a moving carotid artery (Fig 1, E).⁷ Carotid ultrasonography revealed that the LCCA moved to and from the retropharyngeal position with swallowing and was thus being compressed by the hyoid bone (Supplementary Video S1). Corticosteroid therapy was initiated with 30 mg of prednisolone on the 11th hospital day. Her symptoms were immediately relieved and her radiological findings improved (Fig 2, A,B). The inflammatory markers normalized after treatment as well. After tapering down the dosage of prednisolone to 12.5 mg daily, she was discharged on day 51.

Discussion

Arterial thrombosis related to carotidynia is rare. Only 1 case of a patient presenting with amaurosis fugax has been reported.⁶ To the best of our knowledge, this is the first illustration of cerebral embolism due to carotidynia.

Vasculitis in rheumatoid arthritis is an uncommon clinical feature (referred to as rheumatoid vasculitis [RV]). Involvement of RV is usually found in medium- and small-vessel disease, and the skin, digits, peripheral nerves, eyes, and sometimes heart are commonly affected.⁸ On the contrary, involvement of RV in large-vessel disease is rare and central nervous system manifestations, including stroke, are unusual. Although the precise relationship between

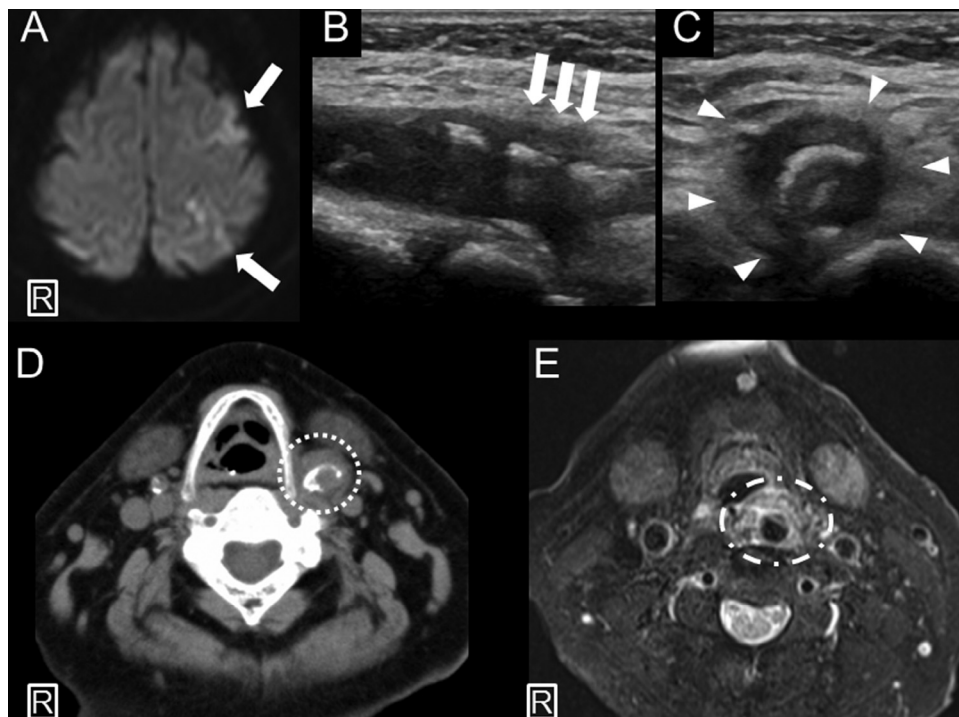


Figure 1. Diffusion-weighted brain magnetic resonance imaging (MRI) revealing high-intensity areas in the left frontal and parietal lobes (arrows) (A). Longitudinal B-mode carotid ultrasonography demonstrating a calcified plaque with swelling of the vessel wall at the bifurcation of the left common carotid artery (LCCA) (arrows) (B). Transverse B-mode imaging showing hypoechoic soft tissue surrounding the area (arrowheads) (C). Noncontrast cervical computed tomography showing isodense tissue surrounding the bifurcation of the LCCA (dotted circle) (D). Fat-saturated T2-weighted cervical MRI revealing high-intensity areas around the bifurcation of the LCCA that is transposed to a retropharyngeal location (dashed circle) (E).

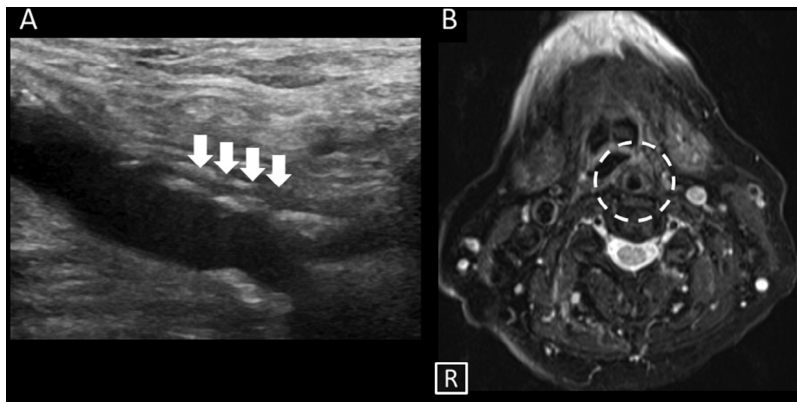


Figure 2. Carotid ultrasonography and cervical magnetic resonance imaging (MRI) after steroid treatment. Longitudinal B-mode carotid ultrasonography revealing decreased swelling of the vessel wall at the bifurcation of the left common carotid artery (LCCA) (arrows) (A). Fat-saturated T2-weighted cervical MRI showing fewer high-intensity areas around the bifurcation of the LCCA (dashed circle) (B).

the medical history of rheumatoid arthritis and carotidynia in our case remains unclear, the clinical presentation of our patient was not consistent with that of typical RV.

Takayasu arteritis and giant cell arteritis are the most prevalent types of large-vessel vasculitis (LVV). Vasculitis of the aortic branches is common in LVV. Arterial involvement in these diseases is usually contiguous in the aorta and often symmetric in paired branch vessels.⁹ As the pathological features suggest, the inflammation of LVV commences from the adventitia and progresses to the intima, resulting in stenosis, occlusion, dilatation, and sometimes aneurysm formation.¹⁰ Luminal narrowing and occlusion in LVV are recognized as a “halo sign” or “macaroni sign” on ultrasonography,^{11,12} and long tapered-type stenotic lesions are frequent.¹³ On the contrary, the inflammation of carotidynia is primarily in the adventitia and surrounding soft tissue,¹⁴ and the lesion is restricted at the carotid bifurcation. The radiological and ultrasonographic findings in our case were consistent with those of carotidynia.

The significant calcification of the carotid artery suggests that the present patient originally had atherosclerosis. Unfortunately, however, there were no images of the carotid artery before the onset. The most distinctive feature in our case is that the affected LCCA was transposed to and from its retropharyngeal location. This phenomenon is referred to as a “moving carotid artery.”⁷ In our case, repetitive compressions of the carotid arteries by the hyoid bone, recognized as a cause of embolic strokes,^{15,16} were observed on ultrasonography during dynamic transposition of the LCCA with swallowing. The repetitive compressions by the hyoid bone were presumed to provoke shear stress¹⁷ and inflammation of the carotid vessel wall,¹⁵ which was aggravated by discontinuation of steroid therapy in our case. These mechanical and inflammatory stresses might cause dysfunction of endothelial cells, hypercoagulation, platelet hyperaggregation, and vulnerability and rupture of carotid plaques, and may subsequently result in embolic strokes.

Conclusion

We report a case of cerebral embolism due to carotidynia, potentially associated with a moving carotid artery caused by swallowing. Clinicians should note that carotidynia can be an important cause of embolic strokes.

Appendix: Supplementary Material

Supplementary data to this article can be found online at [doi:10.1016/j.jstrokecerebrovasdis.2017.10.008](https://doi.org/10.1016/j.jstrokecerebrovasdis.2017.10.008).

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